REPAIR OF CONGENITAL TRACHEAL STENOSIS WITH A FREE TRACHEAL AUTOGRAFT

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Objectives: **Evaluate the results of a technique for repair of congenital tracheal stenosis by use of a free tracheal autograft.** ***Methods:*** **Between January 1996 and July 1997, six infants with congenital tracheal stenosis resulting from complete tracheal rings underwent repair with a free tracheal autograft. Mean age at the time of repair was 4.9 months; mean weight was 5.4 kg. The approach was through a median sternotomy with cardiopulmonary bypass for respiratory support. The trachea was incised anteriorly through the area of stenosis, the midportion of the stenotic trachea was excised, and an end-to-end anastomosis was carried out posteriorly. The excised tracheal segment (1.3 to 2.2 cm long) was used as a free autograft to patch the lower trachea anteriorly. In four infants the autograft was augmented in the upper trachea with pericardium; in two patients with a shorter length of stenosis, the autograft completed the repair. Simultaneous pulmonary artery sling repair (4), ligation and division of patent ductus arteriosus (3), cricoid split (2), atrial and ventricular septal defect repair (1), and complete atrioventricular canal repair (1) were performed at the time of tracheal repair.** ***Results:*** **The infants were extubated and discharged at a mean of 13 and 23 days postoperatively, respectively. One infant had recurrent tracheal stenosis related to the pericardial patch and required a tracheal stent and tracheostomy 4 months postoperatively. Our mean follow-up is 11 months. Bronchoscopic findings currently show widely patent tracheal lumina in all infants.** ***Conclusions:*** **The technique of free tracheal autograft with and without pericardial augmentation was successful in opening the airway of six infants with congenital tracheal stenosis and is currently our procedure of choice for children with this diagnosis. (J Thorac Cardiovasc Surg 1998;115:869-74)**

Surgical alternatives for infants with congenital tracheal stenosis resulting from complete tracheal rings include pericardial tracheoplasty,[1-3] cartilage tracheoplasty,[4] slide tracheoplasty,[5] resection with end-to-end anastomosis,[6] and the recently introduced tracheal homograft.[7] Our experience with infants during and after pericardial patch and slide tracheoplasty combined with recent reports of tracheal homograft transplantation led us to attempt the use of a resected portion of the patient's own trachea as an anterior tracheal patch—a free tracheal autograft.

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Patients and methods

The study included six consecutive infants with tracheal stenosis as a result of complete tracheal rings who were treated with the tracheal autograft technique between January 1996 and July 1997 (Table I). There were three boys and three girls. Age ranged from 10 days to 12 months (mean age 4.9 months). Weight ranged from 2.9 to 10 kg (mean 5.4 kg). Symptoms included respiratory distress requiring intubation and emergency transfer (3), severe stridor with cyanotic episodes (2), and difficult intubation at the time of atrioventricular canal repair (1). In all infants the diagnosis was confirmed by examination with a rigid bronchoscope. In many cases the bronchoscope itself could not be passed through the rings, but the

Table I. *Clinical characteristics*

Patient No.	*Age (mo)*	*Weight (kg)*	*Associated anomalies*	*No. of complete tracheal rings*	*Autograft length (cm)*	*Pericardial patch length (cm)*	*Intubation (days)*	*Discharge (days)*	*Follow-up (mo)*
1	2	4.5	PA sling, VSD, left SVC, ASD, tracheal RUL	15	1.5	2.5	13	20	20
2	5	4.9	Subglottic stenosis, laryngomalacia	6	1.3	—	7	14	14
3	2	4.2	PA sling, PDA, hypoplastic right lung	16	1.3	3.0	7	19*	12
4	0.3	2.9	PA sling, subglottic stenosis, left SVC, PDA	18	2.2	1.5	21	34	10
5	12	10	PA sling, hypoplastic right lung	20	2.0	3.0	15	28	5
6	8	6.2	CAVC, PDA	8	1.8	—	14	21	2
Mean	4.9	5.4		14	1.7	2.5	13	23	11

ASD, Atrial septal defect; *CAVC,* complete atrioventricular canal defect; *PA,* pulmonary artery; *PDA,* patent ductus arteriosus; *RUL,* right upper lobe; *SVC,* superior vena cava; *VSD,* ventricular septal defect.

*Required multiple readmissions over a 5-month period.

fine telescope (outer diameter = 2.5 mm) was passed to the carina. The number of complete tracheal rings were 15, 6, 16, 18, 20, and 8 rings. The actual length of stenosis was slightly longer (but difficult to precisely measure) because generally a funnel-like effect is present at each end of the complete rings. The tracheal lumen at the narrowest site was 2.5, 2.5, 2.0, 2.5, 3.0, and 2.5 mm. All patients were evaluated for pulmonary artery sling and other cardiac anomalies by cardiac echocardiography.[8] Associated anomalies included pulmonary artery sling (4), patent ductus arteriosus (3), subglottic stenosis (2), left superior vena cava (2), hypoplastic right lung (2), Down syndrome (2), ventricular septal defect and atrial septal defect (1), complete atrioventricular canal defect (1), and tracheal right upper lobe bronchus (1).

Surgical technique. All patients were operated on through a median sternotomy with the use of hypothermic (32° C) cardiopulmonary bypass. In the two patients with congenital cardiac anomalies (atrial septal defect, ventricular septal defect, complete atrioventricular canal defect), complete intracardiac repair was performed first using cardioplegia. In three patients the pulmonary artery sling was repaired by transecting the left pulmonary artery and anastomosing it to the main pulmonary artery anterior to the trachea.[9] In one patient with pulmonary artery sling, hypoplastic right lung, and a tiny right pulmonary artery, the left pulmonary artery was translocated anterior to the trachea when the trachea was divided, without a vascular anastomosis.[10]

While the patient was on cardiopulmonary bypass, the extent of the tracheal stenosis was reevaluated with a rigid bronchoscope. The area of the tracheal stenosis, as localized bronchoscopically and confirmed by a fine needle passed through the tracheal wall from the operative field, was then opened anteriorly in the midline. If the bronchoscope or telescope could not be passed preoperatively, the trachea was inspected after the anterior tracheal incision. In four patients this incision extended from just below the cricoid to the carina (Fig. 1). The midportion of the tracheal stenosis was then excised and used as a free tracheal autograft. When present, a tracheal right upper lobe bronchus was carefully avoided during both the anterior tracheal incision and during the autograft harvest, as shown in Fig. 1. The proximal and distal tracheal segments were mobilized circumferentially to the cricoid superiorly and to the carina inferiorly. Bilateral hilar releases and freeing of the pulmonary vessels from their pericardial attachments was performed as originally described by Grillo.[11] The trachea was then reapproximated posteriorly with interrupted 6-0 Vicryl (4) or 6-0 PDS (2) sutures (Ethicon, Inc., Somerville, N.J.). Although not shown in the illustrations, an attempt was made to keep the suture *out* of the tracheal lumen. The free tracheal autograft was then used to patch the lower trachea (Fig. 2). The corners of the autograft were trimmed, and the tracheal autograft was anchored to the lower trachea with 6-0 Vicryl or PDS sutures. The remaining opening in the trachea was closed with a patch of pericardium. Three small hemaclips were positioned in the soft tissue adjacent to the carina, the upper aspect of the autograft, and the upper extent of the pericardial patch for radiographic analysis of the position of the endotracheal tube in relation to the autograft. We position the tip of the endotracheal tube at a site that is in the midportion of the autograft. This stents the pericardial patch open without irritating the carina.

The length of the autograft ranged from 1.3 to 2.2 cm, and the length of the pericardium from 1.5 to 3.0 cm (Table I). The length of the tracheal resection was based on the principles originally established by Grillo[11] and our intraoperative assessment of an acceptable tension for the anastomosis. As we obtained experience with the technique, the average length of the autograft was extended from 1.4 cm (first three patients) to 2 cm (last three patients). In one patient (patient 2) with subglottic stenosis and a localized tracheal stenosis (six rings midtrachea), the incision in the anterior trachea measured 2.5 cm long; 1.3 cm of trachea (six rings) was excised as the autograft

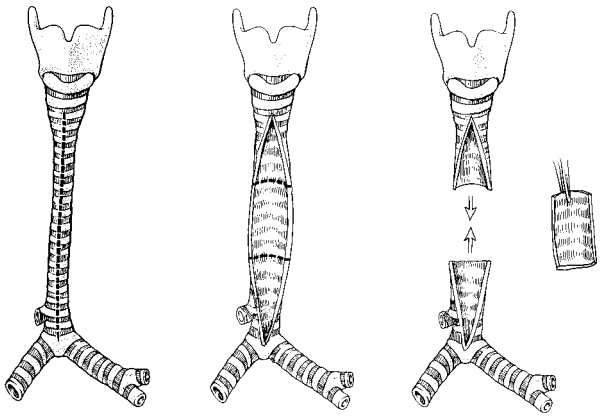

Fig. 1. Patient 1: After median sternotomy and institution of cardiopulmonary bypass, the anterior trachea was incised under bronchoscopic guidance. The trachea was opened from the third tracheal ring to the carina. A portion of the midtrachea measuring six rings long (1.5 cm) was resected to be used as a free autograft. Note the tracheal right upper lobe bronchus that was carefully avoided in both the anterior tracheal incision and the autograft harvest.

(Fig. 3). This particular patient is the only one in the series who might have been a candidate for simple tracheal resection and end-to-end anastomosis, but in our opinion this would have resulted in excessive tension (2.5 cm resection required equaled 50% of the trachea length). A separate incision was made for the cricoid split. The cut ends of the trachea were reapproximated and, after both ends were trimmed, the autograft was inserted, completely closing the residual tracheal opening (Fig. 4). The area of the cricoid split was patched with pericardium because it was readily available and provided an airtight seal.

Results

All patients survived the operation and are currently thriving at 2 to 20 months (mean 11 months) postoperatively. The patients were all extubated between 7 and 21 days postoperatively (mean 13 days). We have used the endotracheal tube as an active stent for the pericardial portion of the repair and prefer for that reason to leave the tube in place for 1 to 2 weeks. The endotracheal tube does not seem to cause much granulation tissue unless the tip irritates the carina. That is a clear advantage of the autograft technique in that the distal trachea adjacent to the carina, where the autograft is positioned, does not require active stenting. Patients were discharged from the hospital 14 to 34 days postoperatively (mean 23 days). Five of the six patients have had essentially no postoperative airway problems. Elective bronchoscopic examinations were scheduled at 1 week postoperatively, just before extubation (≅2 weeks), just before hospital discharge (≅3 weeks), and at 3 and 6 months postoperatively. The third patient in the series with the longest proportional length of pericardium (3.0 cm) had recurrent tracheal stenosis develop in the portion of the trachea patched with pericardium. She also had a hypoplastic right lung. After failure of multiple bronchoscopic dilatations of a recurrent stenosis related to the pericardial patch, a wire expandable Palmaz tracheal stent (Johnson & Johnson Interventional Systems Co., Warren, N.J.) was placed 3 months postoperatively.[12] Because of a failure to clear her secretions, tracheostomy was performed at 4 months postoperatively. This required intermittent hospitalization for a 5-month period. She is currently doing well with her tracheostomy and is 1 year postoperative.

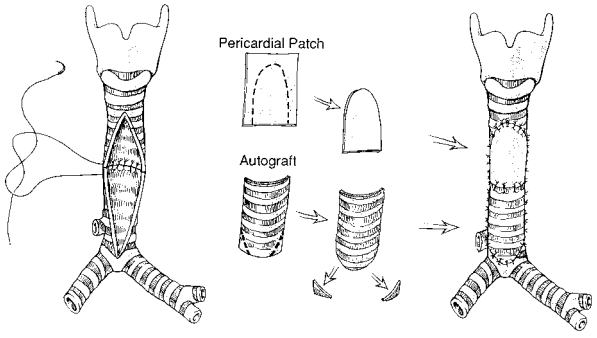

Fig. 2. Patient 1: The two cut ends of the trachea were brought together posteriorly without tension with interrupted sutures. The corners of the lower part of the autograft are trimmed to create a rounded end to fit the trachea above the carina. The lower trachea has been patched open with the tracheal autograft. The upper trachea has been opened with a short pericardial patch (2.5 cm).

Follow-up lung perfusion scan in the patients undergoing pulmonary artery sling repair has demonstrated a percentage blood flow to the left lung of 64% and 77% (translocation technique) in the patients with a hypoplastic right lung; 46% and 24% in the other two patients.

Discussion

We report the cases of six infants with congenital tracheal stenosis from complete tracheal rings who underwent repair using partial tracheal resection, posterior end-to-end anastomosis, and anterior patching of the stenotic trachea with the opened resected trachea, a free tracheal autograft. In four

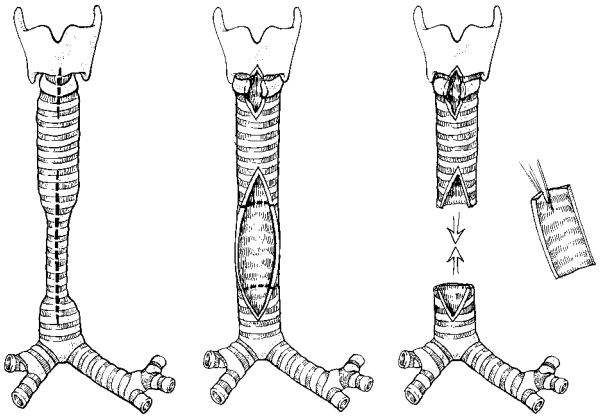

Fig. 3. Patient 2: The lower trachea was incised anteriorly through the extent of the tracheal rings. The area of complete tracheal rings was excised for use as the free tracheal autograft (1.3 cm). The thyroid cartilage, cricothyroid membrane, cricoid, and first two tracheal rings were incised for the cricoid split.

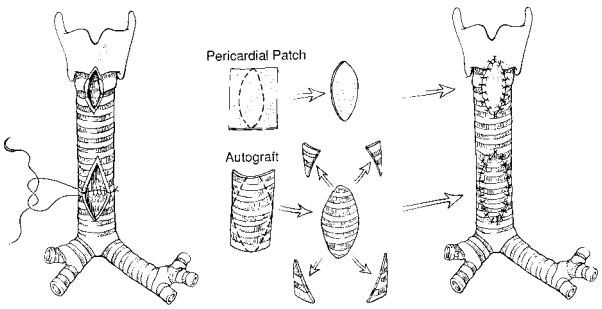

Fig. 4. Patient 2: The cut ends of the trachea were anastomosed posteriorly. The corners of the autograft were trimmed. The anterior tracheal opening was patched completely with the free tracheal autograft. A small pericardial patch was used to seal the area of the cricoid split.

patients the autograft patch was augmented with pericardium superiorly. In two patients, the autograft was long enough to patch the trachea by itself. Patients were discharged a mean of 23 days postoperatively. One patient required a tracheostomy 4 months postoperatively. Currently all patients have completely healed airways and are asymptomatic. Five of six children in this series had risk factors that we have identified as placing them at increased risk for reoperation or stent placement after pericardial patch tracheoplasty.[13] These risk factors include associated pulmonary artery sling (four of six in this series), age less than 6 months (four of six in this series), and tracheal right upper lobe bronchus (one of six in this series). None of these six patients have required reoperation; only one required a temporary stent and still has a tracheostomy.

The conceptual progression that led to our use of the free tracheal autograft was initially based on our observation that the trachea in children with complete tracheal rings is frequently longer than normal. This observation resulted from a cumulative experience at our institution of more than 40 patients with complete tracheal rings. Because of this, pericardial tracheoplasty may result in an excessively long patch that does not have intrinsic support. We surmised that these patients might have an improved outcome after pericardial patch tracheoplasty if the trachea were shorter. In one recent case (November 1995) we therefore excised a 1.5 cm portion of the trachea and reanastomosed the trachea posteriorly before anterior pericardial patch tracheoplasty. Shortly thereafter, we became aware of the recent efforts in Europe using cryopreserved tracheal homograft for infants with complete tracheal rings.[14] We were struck by the fact that the portion of trachea we had excised and sent to the pathology laboratory looked just like the illustrations of a tracheal homograft. The next patient we operated with complete tracheal rings had a very long and severely stenotic lower trachea. After opening the trachea anteriorly, we resected the midportion of the trachea to shorten the resulting pericardial patch and then decided to use it as an anterior tracheal patch analogous to the technique of tracheal homograft: a free tracheal autograft. In that first patient, the autograft was not long enough to cover the entire tracheal opening; a pericardial patch was used for the upper trachea. In the second patient in this series with a shorter length of stenosis, the autograft was used to completely cover the anterior tracheal opening.

The use of a free tracheal autograft has several advantages over tracheal homograft, slide tracheoplasty, and pericardial tracheoplasty. A tracheal autograft is living tissue already lined with respiratory epithelium, unlike cryopreserved tracheal homograft. It is readily available and not subject to rejection. It has cartilage that intrinsically maintains its contour without a silicone stent as described for tracheal homografts. Unlike tracheal homograft, it has the potential for growth. The free tracheal autograft is technically easier to perform than slide tracheoplasty. The trachea is simply incised anteriorly the length of the tracheal stenosis under bron-

choscopic guidance. Once the trachea is opened, measurements can be made to assess the length of trachea to be resected and used as an anterior autograft. In contrast, slide tracheoplasty requires bronchoscopic assessment of where the precise midportion of the stenosis is located so the trachea can be initially transected in the proper location. In addition, when the slide tracheoplasty is started, the upper trachea has to be lifted anteriorly for an initial series of posterior sutures that are difficult to see and tie, especially in a small infant. All the suturing with the autograft is anterior except for the end-to-end anastomosis, which is easily visualized through the anterior tracheal opening. The autograft technique has a technical advantage over pericardial tracheoplasty in that the airway is stable from the time of surgery, obviating the necessity for prolonged endotracheal intubation or later stent placement. Of course, simple resection with end-to-end anastomosis is ideal for shorter lengths of tracheal stenosis. However, in our total experience of 40 infants with tracheal stenosis, only two were candidates for simple resection. It should be noted that the length of "stenosis" is always longer than the actual length of the complete tracheal rings because of the funnel-type effect at both ends of the complete rings. An advantage of the autograft technique over an extensive resection is that none of the suture lines are under tension, avoiding the complication described by Jacobs and associates.[14]

The autograft appears to heal to the adjacent trachea very quickly. It also heals to the adjacent pericardium. A minimal formation of granulation tissue appears at the anastomotic sites. No autograft necrosis has occurred despite complete devascularization of the autograft. Our original fears with regard to this were tempered by the results of homograft implantation.

Although theoretically the autograft technique could be used to completely patch the trachea in a child with rings from cricoid to carina, we have felt more comfortable with a composite patch of pericardium and autograft. We have recently reported that pericardium ultimately becomes covered by pseudostratified columnar epithelium.[15] Our experience is that a one-third resection of the trachea (1.5 to 2.0 cm, six to eight rings) seems safe, with the posterior tracheal anastomosis under acceptable tension.[16] One patient with a relatively long pericardial patch, patient 3, had recurrent tracheal stenosis develop in the area of the patch and required tracheostomy. The success of the pericardium in the other patients may be related to the relatively short length of pericardium that is then well supported by the surrounding trachea and autograft. In particular, the autograft appears to maintain a patent airway at the critical lower trachea and carina.

Conclusions

Repair of congenital long-segment tracheal stenosis using a free tracheal autograft has been successful in six consecutive infants. In four patients the autograft was augmented superiorly with pericardium. This technique combines several advantages of resection with end-to-end anastomosis along with tracheal homograft and slide tracheoplasty using principles derived from a long experience with pericardial tracheoplasty. Best results in this limited series were achieved with a longer length of autograft and shorter length of pericardium. Longer follow-up is needed to assess tracheal growth, but this is currently our procedure of choice for these infants.

REFERENCES

1. Idriss FS, DeLeon SY, Ilbawi MN, Gerson CR, Tucker GF, Holinger L. Tracheoplasty with pericardial patch for extensive tracheal stenosis in infants and children. J Thorac Cardiovasc Surg 1984;88:527-35.
2. Cosentino CM, Backer CL, Idriss FS, Holinger LD, Gerson CR, Mavroudis C. Pericardial patch tracheoplasty for severe tracheal stenosis in children: intermediate results. J Pediatr Surg 1991;26:879-85.
3. Bando K, Turrentine MW, Sun K, et al. Anterior pericardial tracheoplasty for congenital tracheal stenosis: intermediate to long-term outcomes. Ann Thorac Surg 1996;62:981-9.
4. Jaquiss RDB, Lusk RP, Spray TL, Huddleston CB. Repair of long-segment tracheal stenosis in infancy. J Thorac Cardiovasc Surg 1995;110:1504-12.
5. Grillo HC. Slide tracheoplasty for long-segment congenital tracheal stenosis. Ann Thorac Surg 1994;58:613-21.
6. Nakayama DK, Harrison MR, de Lorimier AA, Brasch RC, Fishman NH. Reconstructive surgery for obstructive lesions of the intrathoracic trachea in infants and small children. J Pediatr Surg 1982;17:854-71.
7. Jacobs JP, Elliott MJ, Haw MP, Bailey CM, Herberhold C. Paediatric tracheal homograft reconstruction: a novel approach to complex tracheal stenoses in children. J Thorac Cardiovasc Surg 1996;112:1549-60.
8. Alboliras ET, Backer CL, Holinger LD, Mavroudis C. Pulmonary artery sling—diagnostic and management strategy. Pediatrics 1996;98(suppl II):530.
9. Backer CL, Idriss FS, Holinger LD, Mavroudis C. Pulmonary artery sling: results of surgical repair in infancy. J Thorac Cardiovasc Surg 1992;103:683-91.
10. Jonas RA, Spevak PJ, McGill T, Castaneda AR. Pulmonary artery sling: primary repair by tracheal resection in infancy. J Thorac Cardiovasc Surg 1989;97:548-50.

11. Grillo HC. Surgical approaches to the trachea. Surg Gynecol Obstet 1969;129:347-52.
12. Filler RM, Forte V, Fraga JC, et al. The use of expandable metallic airway stents for tracheobronchial obstruction in children. J Pediatr Surg 1995;30:1050-6.
13. Backer CL, Mavroudis C, Dunham ME, Holinger LD. Reoperation after pericardial patch tracheoplasty. J Pediatr Surg 1997;32:1108-12.
14. Jacobs JP, Haw MP, Motbey JA, Bailey CM, Herberhold C, Elliott MJ. Successful complete tracheal resection in a three-month-old infant. Ann Thorac Surg 1996;61: 1824-7.
15. Cheng ATL, Backer CL, Holinger LD, Mavroudis C, Dunham ME, Gonzalez Crussi F. Histopathological changes following pericardial patch tracheoplasty. Presented at the 12th Annual Meeting of the American Society of Pediatric Otolaryngology, Scottsdale, AZ, May 14, 1997.
16. Backer CL. Successful complete tracheal resection in a three month infant [invited commentary]. Ann Thorac Surg 1996; 61:1824-7.